

Selective reduction of a triplet heterotopic cervical pregnancy after embryo transfer

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ABSTRACT

Cervical pregnancy is rare and heterotopic cervical pregnancy is more uncommon ever. Assisted Reproduction Technology enhances these pregnancies. The diagnosis and the treatment which are in emergency can be sometimes difficult. This article is a case report of a triplet heterotopic cervical pregnancy after embryo transfer, and a scientist review of the literature found in MEDLINE. To date, in the English literature, we found only 7 cases report of triplet heterotopic cervical pregnancies.

Keywords: Heterotopic Pregnancy; Cervical Pregnancy; Ectopic Pregnancy; IVF; Embryo Transfer; Assisted Reproduction Technology

1. INTRODUCTION

Heterotopic pregnancy is an association between an intrauterine and an ectopic pregnancy (tubal or more rarely ovarian, cervical, abdominal). The first description comes from Duvernay in 1761 during an autopsy [1]. It is a rare natural phenomenon (less than 1/30000 pregnancies) [2,3], but its rate increases with the introduction of Assisted Reproductive Technology (ART) (0.8% - 3% after IVF) [4,5].

Cervical pregnancies are very rare (1/2500 - 18000 pregnancy and <1% ectopic pregnancies) [6-9] and heterotopic cervical pregnancies, usually resulting from ART, are even more rare [10].

This is the first report of an ongoing pregnancy after a selective aspiration of the cardiac fluid of a triplet heterotopic cervical pregnancy.

2. CASE REPORT

Our patient was a 36 years old, with 3 previous spontaneous conceptions and deliveries, with an another partner. She underwent a tubal ligation after her last delivery. A few years later, she consulted with a new partner. Her partner had a normal sperm count. So she underwent a tubal reanastomosis which was unsuccessful with a bilateral tubal obstruction seen on her post operative hysterosalpingography. After discussing the alternatives, the couple decided to undergo IVF.

The patient received a long agonist protocol using a gonadotrophin-releasing hormone (GnRH) agonist (Suprefact; Sanofi-Aventis) introduced in the luteal phase of the previous cycle at a dose of 0.5 mg SC daily and then decreased to 0.25 mg daily at the initiation of gonadotrophin stimulation. The patient received 225 IU daily of hMG (MENOPUR; Ferring, Canada) that was increased to 300 IU in association with 150 IU of recombinant FSH (GONAL-F; EMD Serono Canada). Final oocyte maturation was triggered by 10000 IU human chorionic gonadotrophin (Pregnyl, Merck, Canada). The patient underwent oocyte retrieval 36 hours after receiving hCG; 14 oocytes were retrieved. After insemination with sperm, we obtained 9 embryos. The patient had an ultrasound guided embryo transfer of 3 day3 embryos (8C2-3, 8C2-3 and 10C2). The transfer technique was described as easy by the physician. Two weeks later, hCG level was 186.8 UI/L and 331.5 UI/L two days after.

At 8 weeks and 3 days of amenorrhea, a trichorionic triamniotic triplet pregnancy with one gestationnal sac in the cervix was discovered at endovaginal ultrasound. The 3 embryos had a positive heart beat.

After discussing the situation with the couple, they decided to undergo a embryonic reduction of the cervical

pregnancy. They understood and accepted the procedure, including the risk of bleeding and the risk of losing the three embryos.

At 8 weeks and 4 days of amenorrhea, in the operating room, the patient had para cervical block with XYLOCAINE 1%. We used the 7.5 MHZ endovaginal ultrasound probe (Voluson E8, GE, Healthcare, Baie d'Urfé, QC, Canada) with a needle guide. Using an egg collection needle (OPS 30cm, 19G, CCD, France), we aspirated the amniotic fluid and then the cardiac blood of the cervical embryo. No heart beat was seen for the cervical embryo at the end of the procedure, and the intrauterine embryos had normal heart beat. The procedure was easy and the patient did not have any pain or bleeding.

A 12 weeks ultrasound showed a dichorionic diamniotic ongoing twin pregnancy

The patient had a normal pregnancy that was complicated by premature labor, an emergency C-section was performed at 25 weeks; she delivered a girl (660 g) and a boy (620 g).

3. DISCUSSION

Heterotopic cervical pregnancy is a very rare event. ART use has increased the rate of heterotopic pregnancies with unusual localisation like cervix or cesarean section scar [11,12].

The diagnosis of a cervical pregnancy can be difficult and delayed [13-15]. The main symptoms are bleeding during the first trimester, abnormal hCG increase. The transvaginal ultrasound is usually diagnostic (the first report has been described by Raskin in 1978) [16] but magnetic resonance imaging (MRI) has been necessary in some cases to make the definitive diagnosis [17,18]. The diagnosis is confirmed by finding an empty uterine cavity, in association with a gestational sac with an embryo (with a positive or negative heart beat) in the endocervical canal below the internal os level. A heterotopic cervical pregnancy can be even more difficult to diagnose, because someone can focus on the intrauterine pregnancy without seeing the cervical one.

Cervical pregnancy is a therapeutic emergency because of the risk of severe hemorrhage. There were even recent reports of death following a heterotopic cervical pregnancy [19].

Different treatments for ectopic cervical pregnancy are described in the literature: systemic Methotrexate injection [20-22], uterine artery embolization or ligation [23] [24], transvaginal ultrasound-guided aspiration [23], KCl solution [25], Methotrexate [13,26,27] or hyperosmolar glucose injection into the gestational sac [28], cervical curettage with balloon tamponade [29-31] or Shirodkar cerclage to make hemostasis [32]. Olav describes a hysteroscopy resection of a cervical pregnancy because of bleeding after Methotrexate treatment [33].

There is no consensus on the technique to use for treatment. Prognostic factors that affects the effectiveness of the conservative treatment are especially the age of pregnancy [34,35]. Sometimes, hysterectomy is inevitable because of severe bleeding [13,36,37].

In our case, systemic Methotrexate and uterine artery embolization or ligation were not an option because of the presence of an ongoing intrauterine pregnancy.

We chose the selective reduction by the fetal heart aspiration technique, but Sijanovic *et al.* tried selective Methotrexate injection, and had a good result [26]. Some articles describe the injection of KCl in fetal heart or gestational sac [19,38-46]. Some authors injected Actinomycin D [47]. Jozwiak *et al.* resected the cervical pregnancy by hysteroscopy and coagulated the bleeding sites with roller-ball. The remaining intra uterine pregnancy then continued successfully to term [48]. Kim *et al.* and Vasiliki *et al.* aspirated the cervical pregnancy and achieved hemostasis by compression with pediatric Foley catheter [30,49].

All these techniques have three goals: minimize the bleeding, preserve the fertility and the intrauterine pregnancy. We have to keep in mind that if these conservative treatments fail or if the patient starts to bleed severely, there would no choice but to sacrifice the intrauterine pregnancy with the first treatments methods described.

The mechanism to explain a cervical pregnancy is unclear. Cervical or uterine surgeries in the past (curettage or cesarean), seem to enhance these ectopic pregnancies [36,37,50,51]. Does the problem come from an unreceptive endometrium?

For the rare spontaneous cervical pregnancies, some authors wonder if the transport of the ovum would be too quick and that the embryo arrives in the uterine cavity with an unprepared endometrium [52,53].

A chromosomal abnormality in the embryo is another hypothesis [51,54].

As mentioned before, these ectopic pregnancies are more common after ART and specially after IVF [40,48, 55-58]. Furthermore, in the english literature, we found only 7 cases report of triplet heterotopic cervical pregnancies (6 with intrauterine twins and one with cervical twins) [5,28,44,59-62]. None of them were spontaneous, all these pregnancies were the result of embryo transfer after IVF. So we can say that IVF and transferring more than one embryo are risk factors for heterotopic pregnancies.

4. CONCLUSION

Historically, many women with a cervical ectopic or heterotopic cervical pregnancy underwent hysterectomy. With the development of transvaginal ultrasound, the diagnosis of these rare pregnancies has become easier and quicker. A conservative method should be tried first,

especially as these pregnancies are in infertile patients undergoing ART. This is the first report of an ongoing pregnancy after a selective aspiration of the cardiac fluid of a triplet heterotopic cervical pregnancy.

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